

## Untreatable Pelvic Pain in A Young Woman for Aneurysmal Bone Cyst: A Description of a Rapid Solution with a Selective Embolization

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### Abstract

**Introduction:** Aneurysmal bone cyst (ABC) is a benign, rare, highly vascularized, and relapsed tumor. It has a locally aggressive behavior characterized by rapid growth. Surgical resection is considered the first-choice treatment for ABC. However, as pelvic lesions are complex to manage surgically, other less invasive treatments have been introduced, such as selective arterial embolization (SAE). In this work we expose a case of ABC with in-depth analysis of the diagnostic process, the percutaneous treatment, and the clinical and radiographic results.

**Case presentation:** A young, 20-year-old woman in good health comes to visit for persistent pain in the left groin, which began over 1 year ago and has increased in recent months, constantly, with also functional limitation to active and passive mobilization of the non-responsive left hip to pharmacological treatment. Instrumental tests are performed for further diagnostic analysis of the reported symptoms. The MRI of the pelvis shows a voluminous osteo-destructive formation (DT max 8 cm), with serum-blood content and contextual fluid-fluid levels, affecting the left ilio-pubic branch, compatible with an ABC lesion. The patient underwent SAE as primary treatment, without showing any peri- and post-procedural complications. In the subsequent clinical and instrumental checks, the progressive and slow volumetric reduction of the lesion is observed, with the appearance of a periosteal calcification component. The patient reports a progressive decrease in painful symptoms and a return to physical activity.

**Conclusion:** In this case report, we expose the effectiveness of SAE as a first-choice treatment in patients with ABC of the pelvis, a complex site to treat surgically. This procedure guaranteed the complete resolution of the painful symptoms with the absence of peri- and post-procedural complications.

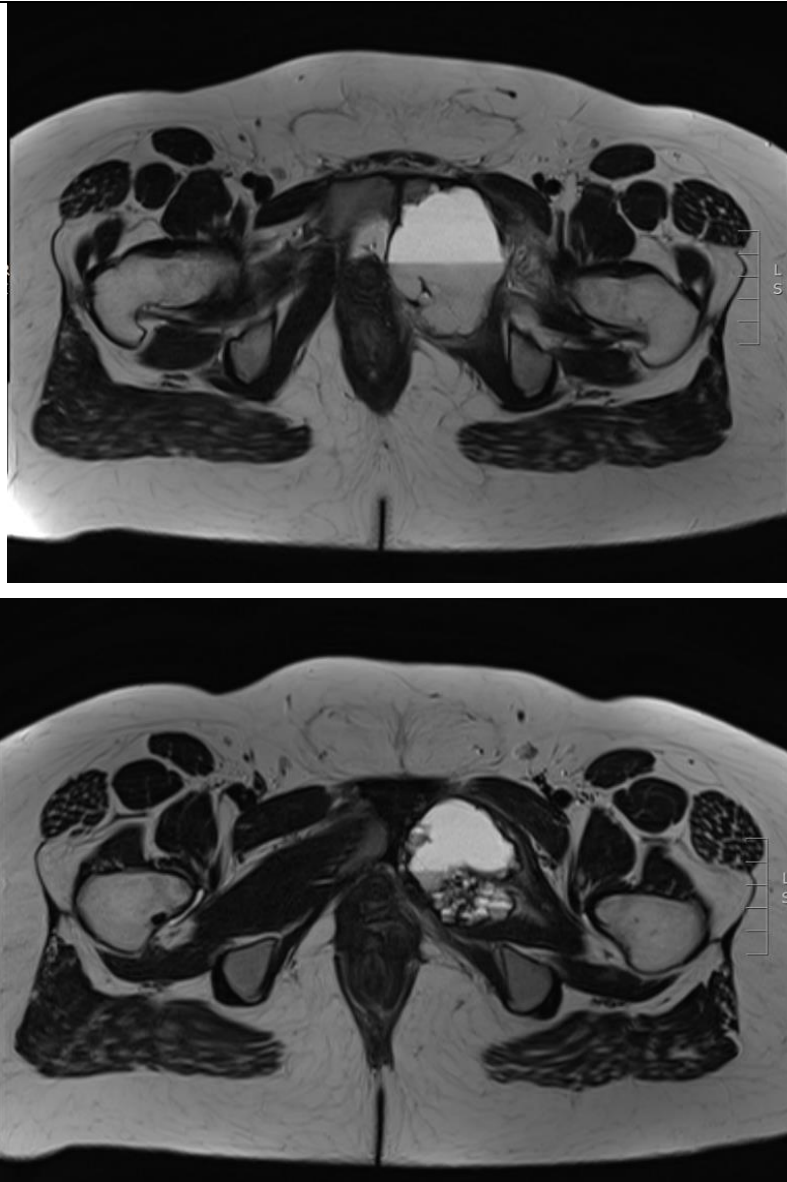
**Keywords:** Cyst; Bone; Aneurysmal; Embolization; CT; Arteriography; Pelvis; MRI; SAE

## Introduction

Aneurysmal Bone Cyst (ABC) is a benign, rare, highly vascular, and relapsed tumor [1]. It has a locally aggressive behavior characterized by rapid growth. ABC represents 1-2% of primary bone tumors, begins more frequently in the first 2 decades of life, and has a slight prevalence in females [2]. In 70% of cases, ABCs appear as primary bone lesions, while in 30% of cases they are secondary to giant cell tumors, chondroblastoma, osteoblastoma and osteosarcoma [3,4]. In the literature a third cause is described. ABC may be secondary to previous trauma [5,6]. ABC is localized in 12% of cases at the pelvis level but can occur in any bone district [7,8]. In 52% of cases, ABC affects the metaphysis of the long bones, sternum, and spinal column [4]. They are lesions of unknown etiology. However, they seem to be associated with malformations of the bone vascular component, of unknown cause. These malformations cause an increase in pressure on the adjacent bone trabeculae, causing progressive erosion and resorption of the surrounding bone structures [5,6]. Clinically, ABC manifests as acute pain with swelling and possible formation of a palpable mass. Over time, the pain becomes chronic lasting from weeks to months. ABC rarely occurs with intense pain in patients with pathological fractures. When present in locations such as the pelvis or spine, the painful symptoms can be associated with neurological disorders of various degrees. These neuropathies are secondary to compression by the ABC of the surrounding nervous structures [6,9]. Surgical resection is considered the first-choice treatment for ABC. However, as pelvic lesions are complex to manage surgically due to the difficult to access location, the large size of these masses and the proximity to noble neurovascular structures [10], other less invasive treatments have been introduced, such as Selective Arterial Embolization (SAE) [11,12]. We expose a case of ABC with in-depth analysis of the diagnostic process, the percutaneous treatment, and the clinical and radiographic results up to the last follow up.

## Case Presentation

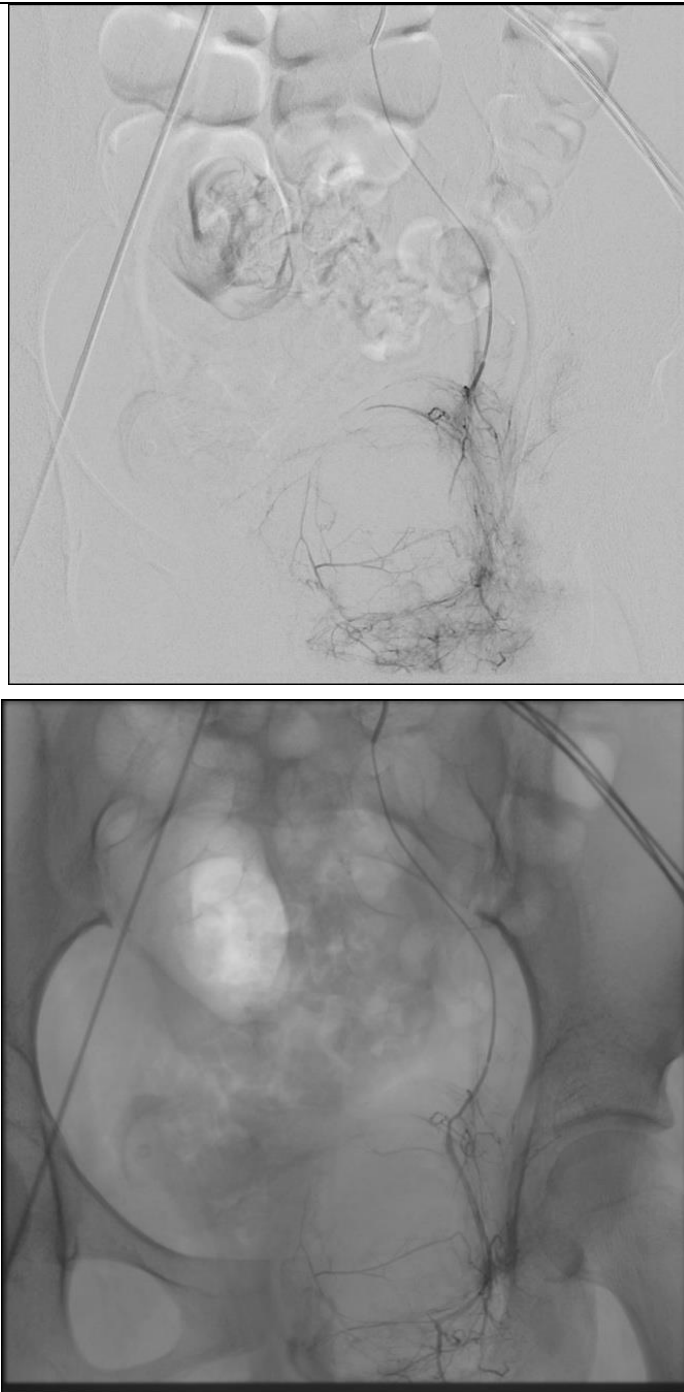
A young, 20-year-old woman in good health comes to a visit for persistent pain in the left groin, which began over 1 year ago and has increased in recent months, now becoming constant, with algo-functional limitation to the active and passive mobilization of the left hip unresponsive to pharmacological therapy. Instrumental tests are performed for further diagnostic analysis of the reported symptoms. The MRI of the pelvis shows a voluminous osteo-destructive formation (DT max 8 cm), with serum-blood content and contextual fluid-fluid levels, affecting the left ilio-pubic branch, compatible with an aneurysmal bone cystic lesion (Figure 1).



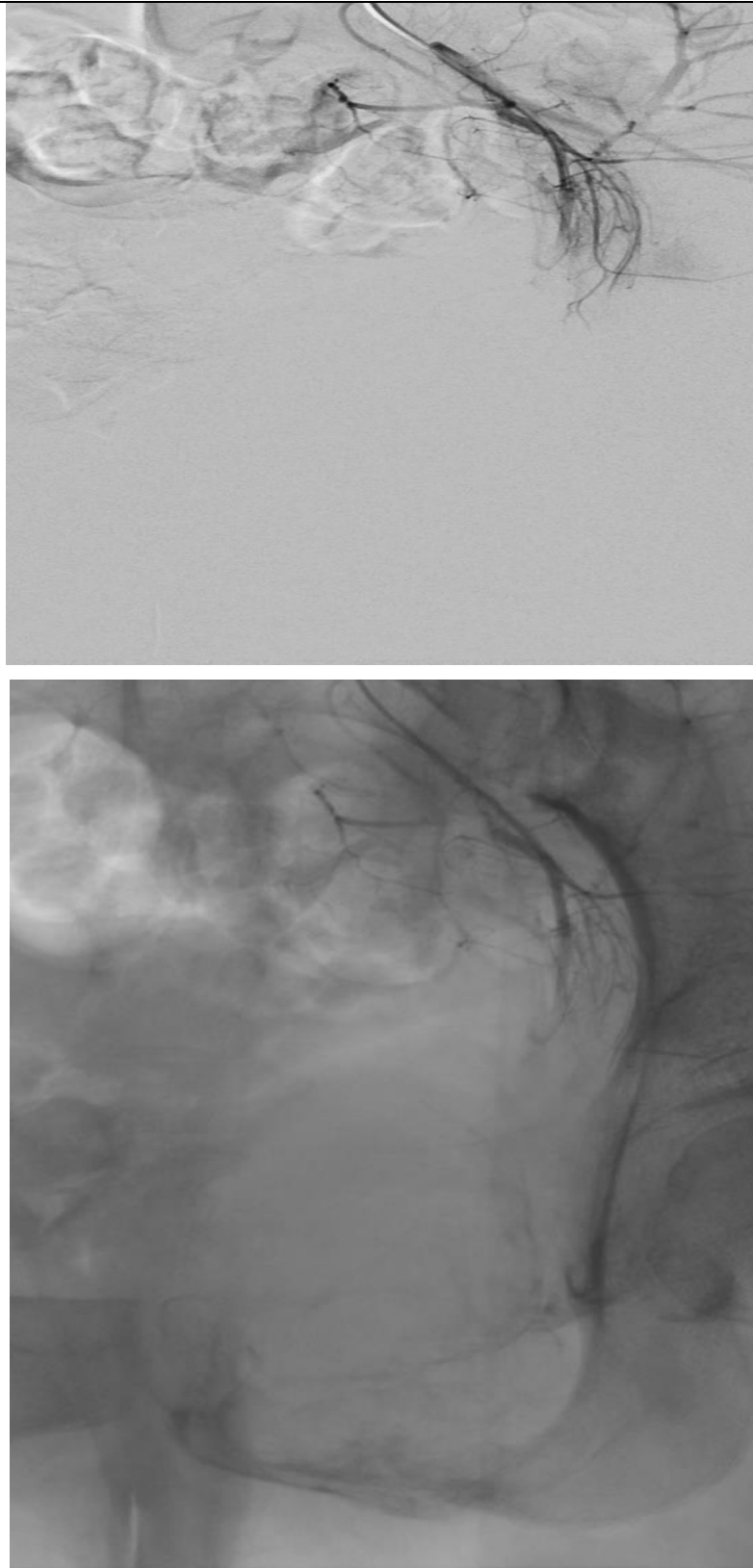
**Figure 1:** MRI axial T2w of pelvis shows a voluminous lesion with serum-blood content and fluid-fluid levels.

The patient was subjected to a questionnaire according to the NPRS pain scale (Numeric Pain Rating Scale), a universal scale to measure patient-reported pain. It consists of a scale ranging from 0 for “no pain” to 10 for “the worst possible pain” [13,14]. The NPRS result is a score of 9, corresponding to worsening pain. Following further diagnostic studies, diagnostic angiography and subsequent SAE of the affected arterial branches are planned. The patient gives informed written consent to the procedure. Ultrasound-guided retrograde right femoral access is performed, and a 5 French (Fr) diameter introducer is positioned. After selective catheterization of the left hypogastric artery "in crosse", the arteriography highlights an oval area with anomalous peripheral impregnation, corresponding to the known bony lesion of the ipsilateral ilio-pubic branch (Figure 2). The lesion appears supplied by branches of the hypogastric anterior branch. We therefore proceed with 2.7 Fr and 2.0 Fr super selective microcatheterization of the main branches supplying the lesion, originating from the pudendal and obturator arteries. Furthermore, further thin newly formed alien branches are appreciated which are 2.0 Fr super selectively microcatheterized. The aforementioned branches are embolized with 500-700- and 700-1000-micron Contour particles; two particle vials are used in total. The final

arteriographic checks show devascularization of the target area with preserved flow in the adjacent vascular structures (Figure 3). Manual hemostasis of the puncture site is performed for 20' with sterile gauze and positioning of an adhesive pneumatic device in the right groin. The treatment showed no peri- and post-procedural complications.



**Figure 2:** Arteriography shows an oval area with anomalous peripheral impregnation, corresponding to the bony lesion of the ipsilateral ilio-pubic branch.



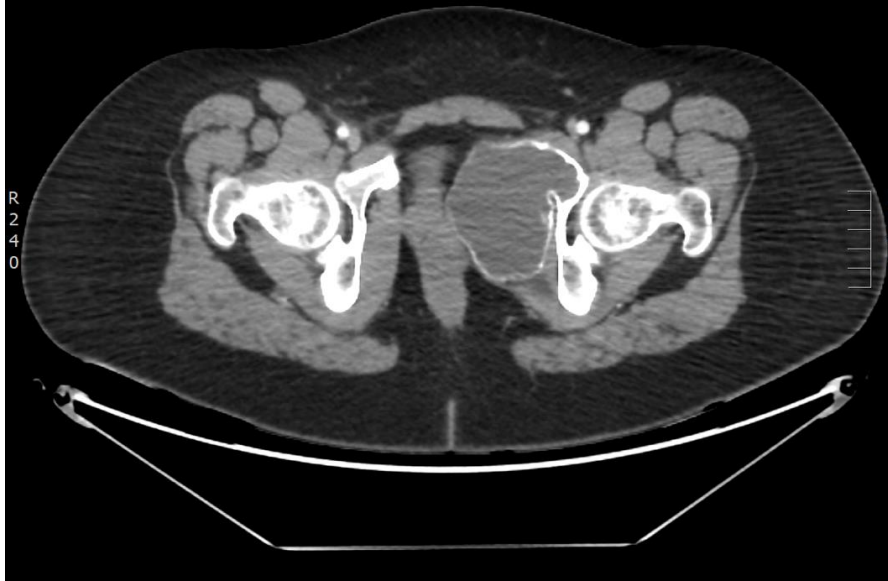
**Figure 3:** The final arteriography shows devascularization of the target area with preserved flow in the adjacent vascular structures.

In subsequent CT checks performed at 1 month (**Figure 4**) and 1 year (**Figure 5**), the progressive and slow volumetric reduction of the lesion is observed, with the appearance of a periosteal calcification component. Progressive

calcification of the walls of the ABC is a slow process, which may take over a year. This feature, however, is not observable in all patients with ABC treated with minimally invasive techniques [15]. Clinically, the patient reports a progressive decrease in painful symptoms, with a 1year NPRS score of 2. In the follow-up by MRI at 6 months and 2 years (Figure 6), the progressive reduction in size of the lesion and disappearance of the fluid-fluid level are confirmed. The patient is subjected to an NPRS questionnaire to evaluate the clinical evolution of painful symptoms two years after treatment. The result of this questionnaire is 0 or absence of pain, reporting progressive loss of pain, functional recovery up to a complete resolution of painful symptoms 2 years after treatment with SAE.



**Figure 4:** CT follow up of the lesion after 1 month.



**Figure 5:** CT follow up 1 year shows a progressive and slow volumetric reduction of the lesion, and the appearance of a periosteal calcification component.





**Figure 6:** MRI of pelvis shows the progressive size reduction of the lesion and disappearance of the fluid-fluid level.

### **Discussion**

In this case report we discuss the use of SAE as a primary treatment for ABC, particularly in a site such as the pelvis, which is more difficult to manage surgically. The ABC is localized in 12% of cases at the pelvis level [7,8]. Surgical resection is considered the first-choice treatment for ABC due to their benign nature, high vascularity and the significant risk of fractures and recurrences associated with it [16]. Mankin et al. [1] they discussed the surgical treatment of ABCs, located in various sites, in a cohort of 150 patients. In their study, patients underwent curettage and autologous or heterologous transplantation of bone tissue or polymethyl methacrylate. The local recurrence rate of these



lesions is observed to be 20% after surgical treatment. Rossi et al. [17] in their study describe a group of 36 patients with ABC treated with SAE. The persistence of the pathology or the development of a recurrence was found in 14 patients. Of these, 9 patients recovered with a second treatment. The remaining 5 patients recovered on the third SAE attempt. In this group, the only complications observed are skin necrosis (2 cases), scapular necrosis (1 case), groin necrosis (1 case) and transient paresis (1 case). These complications did not adversely affect the patient's outcome. This study highlights the possibility of relapses even with this treatment. At the same time, it is possible to subject the same patient to another SAE, with low risk of complications. To date, less invasive techniques have been introduced for the management of these lesions, such as SAE [11,12]. This technique, together with cryotherapy and intralesional infiltration [1,10,11,18], make it possible to overcome the difficulties associated with the surgical management of the resection. The ABCs, localizing in the pelvis, present difficult surgical accessibility, often large dimensions, and proximity to noble neurovascular structures [10]. The diagnosis of ABC occurs through instrumental tests such as traditional radiography in various projections and magnetic resonance imaging [19]. Differential diagnoses include: unicameral bone cyst, osteoblastoma, fibrous dysplasia, non-ossifying fibroma, and telangiectatic osteosarcoma [16,20]. CT represents a valid tool for a more accurate definition of the extent of the lesion [1]. Biopsy and histological examination allow the diagnosis to be confirmed [1,5,18]. ABCs are characterized by vascularization originating from abnormal arterial structures. Angiography is fundamental for identifying the feeding arteries, allowing us to define which arterial branches to treat. With SAE it is possible to embolize the arteries feeding the pathological lesions without affecting the adjacent healthy tissue [17,21,22]. In the literature, some studies such De Cristofari et al. [15] demonstrate that SAE represents a treatment of primary choice in patients with ABC of the pelvis, sacrum, and spine. In these locations surgery can be dangerous due to intraoperative bleeding and the presence of noble structures that can be accidentally damaged during treatment. Embolization stops the growth capacity of the lesions, favoring their regression, until complete healing and resolution of the symptoms associated with ABC. Embolization is a low-cost, minimally invasive and repeatable procedure compared to traditional surgical treatment [17]. Cevolani et al. have defined the effectiveness of SAE in the low complication rate, the reduced hospital stays, the easy repeatability in case of recurrence, the low cost and the rapid resumption of physical activity [23].

## Conclusion

ABC is a benign, rare, highly vascularized, and relapsed tumor. In this case report we expose the effectiveness of SAE as a first-choice treatment in patients with ABC of the pelvis, a complex site to treat surgically. Our patient was discharged the day following the procedure. This procedure guaranteed the complete resolution of the painful symptoms with the absence of peri- and post-procedural complications. The patient was able to resume her daily activities and gradually also physical activity. In our case, the choice of endovascular treatment was also due to the patient's young age. In fact, surgical treatment presents numerous complications such as possible neurovascular lesions and the development of adhesions which, due to the location of the lesion, can create complications for a future pregnancy. Therefore, we believe that SAE has an excellent cost/benefit ratio in the treatment of ABC and that it is a valid and safe therapeutic option, with excellent possibilities of success as well as easily repeatable in the event of any future relapses.

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